Attitudes About the Use of Newborn Dried Blood Spots for Research: A Survey of Underrepresented Parents

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ABSTRACT

OBJECTIVE. Identify the relative importance of factors that impact parents' attitudes towards use of their child's dried newborn blood spots for research purposes.

METHODS. Respondents were parents aged 18 and older with at least one child aged 17 or younger born in Indiana visiting an urban pediatrics clinic. They were asked to rate the acceptability of hypothetical scenarios involving the research use of blood spots. Three pieces of information varied between the scenarios: 1) who would be conducting the research; 2) whether the child's identity would be linked to the spots; and 3) whether and how often the parents' consent would be sought before the research began.

RESULTS. 506 predominantly Black and low-income parents completed the survey. The conjoint analysis model showed good fit (Pearson's $R=0.998$, $p<0.001$). The rank order of factors affecting parents' attitudes was: 1) consent (importance score=64.9), 2) whether the child's identity was linked to the spot (importance score=19.4), 3) affiliation of the researcher using the spots (importance score=14.6). Respondents preferred being asked for their consent each time their children's spots would be used. They preferred that the children's identity not be linked to the spots and that the research is conducted by university researchers, though these issues had less impact on attitudes than consent.

CONCLUSIONS. Parents strongly prefer that consent be sought for each use of their children's spots. These findings have implications for future research and policymaking decisions.

What's New:

Compared to researcher affiliation and identity linkage of a child to a blood spot, consent protocol emerged as the strongest factor impacting Indiana parents' attitudes about using dried blood spots for research. This has implications for biospecimen use policy.

INTRODUCTION

Beginning in the 1960s, routine newborn screening began in the U.S.[1,2] Since 1965, when it first started screening for phenylketonuria (PKU), the Indiana State Department of Health has collected newborn blood spots and now tests them for nearly four dozen conditions[3,4]. Blood spots are collected for each child, and the residual dried blood spots (DBS) are stored for use in the event that positive findings must be confirmed or that screening must be repeated[5]. Residual DBS are stored indefinitely in Indiana and may be used for quality assurance after screening, but are not released for research purposes[6].

DBS are potentially useful in studies of conditions believed to be genetic or acquired in utero, studies of in utero environmental toxin exposure, and other research on biological conditions[7-11]. In Indiana, for example, given the state's strong electronic medical records systems, e.g., the Indiana Network for Patient Care[12], DBS of Indiana-born children could be linked with their electronic medical records to identify early markers of conditions not currently screened for at birth. Moreover, using DBS is feasible and potentially cost-effective compared to collecting fresh blood specimens[10].

Many states in the U.S. do not explicitly regulate the use of DBS for research purposes, and state policies surrounding the retention and use of DBS vary greatly[13]. Litigation focusing on DBS research use in the U.S. has varied in outcome. For example, Texas now has an opt-in policy where parents can both disallow their child's DBS for use in research and insist that the specimens be destroyed (specifically pertaining to spots collected on or after June 1, 2012)[14,15].

Researchers have long investigated attitudes towards children's participation in a variety of research[16-18], and, more specifically, the use of pediatric biospecimens in research[19,20]. Other

work has focused on attitudes towards using DBS in research[21-25]. Consistently-emerging concerns of respondents in previous research involving use of biomaterial include: anonymity/identifiability of the person whose material it is; the researcher using the biospecimen; and whether and how frequently parent consent is sought, among other factors[21-25].

Policymakers have expressed a need for information concerning parental attitudes surrounding the research use of DBS in order to formulate acceptable policies governing their use[26]. However, little is known about the relative weight that parents place on the factors affecting their attitudes. Here we address the relative importance of three key factors in driving parental attitudes, providing needed information for the development of state policy and institutional research practices.

Our hypothesis was that parents differently weight concerns about the use of their children's DBS. These concerns included: 1) whether consent is sought, 2) the affiliation of the researcher using the DBS, and 3) whether their children's identity is linked to the DBS. We chose these factors based on existing work[21-25] and hypothesized that the issue of consent was likely to emerge as the most significant concern.

METHODS

Survey Development

We employed conjoint analysis, a multivariate technique commonly used in marketing research and increasingly in health-related research, including work on attitudes about biobanking[27]. Conjoint analysis is often used to determine respondent preferences when multiple features or attributes impact a decision[28-30]. We chose a full-profile, ratings-based design. The survey items were generated using SPSS 17®[31].

A full-factorial, within-subjects, repeated measures design produces 12 scenarios (3 levels of consent X 2 levels of identity linkage X 2 levels of who is conducting research). However, we were interested in conceptualizing respondents' ratings as relative preferences, and the conjoint analysis approach aligns with this objective. This approach produced eight experimental survey items. We then strengthened the design by including four additional holdout items. Participants' responses to these holdout items are used as a conjoint analysis model validation check. This resulted in a total of 12 orthogonal survey items. We included a 13th duplicate item as an index of intra-respondent reliability as has been done by other investigators [32].

The paper-based survey's 13 hypothetical scenarios followed the same pattern of information delivery: 1) who (academicians or a pharmaceutical company) would be conducting the research using the DBS, 2) whether the child's identity would be linked to his/her DBS, and 3) whether the parent's consent would be sought once for all research (one-time, blanket consent), every time the child's DBS would be used for research, or not at all. Respondents were asked to rate each of the scenarios via an 11-point semantic differential scale, anchored at "Completely Unacceptable" (0) and "Completely Acceptable" (100). There were five different survey versions with the order of the 13 scenarios randomized to minimize order-effects. Figure 1 reflects survey item wording.

[Figure 1 about here.]

Pilot testing of the survey with 15 English speaking adults confirmed that the survey was understandable and clear. The survey and research protocol were exempted from review by the Indiana University Institutional Review Board.

Study Population, Recruitment, and Procedure

Individuals visiting one of three urban public pediatric clinics or one private pediatric clinic in and around the Indianapolis metropolitan area were approached by a research assistant (RA) from a pediatric research network, a university research group with permission to collect data in clinics affiliated with the Indiana University School of Medicine. Recruitment occurred between June 2011 and March 2012. The IRB waived informed consent for this study. The RA first ensured respondent eligibility through brief screening questions. Eligibility criteria included English fluency, 18 years of age or older, and parent or primary caregiver to at least one child 17 years of age or younger born in the state of Indiana. The RA then asked eligible respondents to keep his or her child in mind when completing the survey. For respondents with multiple children 17 or younger born in Indiana, the RA flipped a coin to determine whether the respondent should focus on the oldest or youngest child born in Indiana as the referent child when completing the survey.

The RA explained the study and asked the respondents to complete a few questions, including ages and sexes of children, whether they recalled the referent child receiving a heelstick before leaving the hospital when the baby was born, and whether they had heard of newborn screening. The RA then provided respondents with general information about newborn screening (the “heelstick”) in Indiana, as well as the storage of the DBS. The RA then informed participants they would rate the acceptability of 13 unique scenarios, each with different information that described fictional (“what-if”) research situations that were not actually taking place, followed by some general demographic questions. The RA reiterated that the survey was anonymous and that the scenarios depicted in the survey were not real.

Respondents completed surveys in 12-15 minutes, primarily while in clinic waiting rooms before appointments. Occasionally, respondents and their children were called back for

their appointments before they completed their surveys. When this occurred, respondents were instructed to return the survey to the RA when their appointments finished. Respondents received a $5 gift card for completing the survey.

An RA scanned the completed surveys. Optical character recognition software identified responses, recorded them to a database, and the RA reviewed each survey's scanned responses to ensure data accuracy.

Data Analysis

Survey responses were analyzed using conjoint analysis. Conjoint analysis produces importance scores, denoting which factors most strongly impact preferences/attitudes, and part-worth utility values, denoting respondents' preferences (here, preferences for what are acceptable research situations). Conjoint analysis also produces Pearson's R and Kendall's tau. The Pearson's R coefficient is an indicator of fit between the conjoint analysis model (i.e., predicted responses) and respondents' observed (i.e., actual) responses to the eight experimental survey items. The Kendall's tau coefficient for the four holdout items was generated; this is the coefficient denoting how well the observed (i.e., actual) responses correlate with the predicted responses as produced by the holdout items. This Kendall's tau for holdouts serves as a model validation index. We also report the correlation for the redundant survey items as a reliability check. All analyses were conducted using SPSS 17®[31].

First, we report overall importance scores for the factors, which denote the impact of each factor (researcher affiliation, identity linkage, and consent) upon acceptability ratings. Specifically, these importance scores denote the percentage of respondents' preferences that are explained by the factors we included in our study, and, therefore, sum to 100 (give or take due to rounding).

Next we examined the part-worth utility values, which sum to zero for each factor. While importance scores indicate how strongly a factor drives one's preferences, part-worth utilities convey the preferences associated for the specific attribute levels *within* those factors (e.g., whether the respondent exhibited a preference for university researchers versus drug company researchers using the DBS).

RESULTS

Of the 593 potential respondents approached, 521 (88%) agreed to participate. The 72 (12% of 593) non-participants were ineligible, uninterested, or indicated they did not have time. Of the 521 who agreed to participate, 506 (97%) respondents completed the survey, and 15 (3%) either did not complete it or returned a survey to the RA with missing pages.

Respondents ranged in age from 18-63 with a mean age of 32 years. The referent child about whom the survey was completed ranged in age from newborn to 17 years of age with a mean age of 5.7 years. Referent children were predominantly male (51%). Respondents were predominantly Black or African American, not Hispanic or Latino, female, and were mother to the referent child. The referent child was most likely a Medicaid recipient. Other descriptive demographic data, including political affiliation, income, and religiosity are noted in Table 1.

[Table 1 about here.]

Over three-quarters of respondents (75%) indicated that they recalled their children receiving a heelstick when they were born. However, fewer respondents indicated that they had heard of newborn screening (60%).

We first calculated overall acceptability ratings for each of the 13 scenarios. Table 2 presents the scenarios and their overall acceptability ratings. In the scenario rated as least acceptable the researcher affiliation was a drug company, the child's identity was linked to the

DBS, and consent was not sought (M = 28.6; SD = 36.4). In the scenario rated as most acceptable, the researcher affiliation was a university, the child's identity was not linked to the DBS, and consent was sought for each and every use of the DBS (M = 78; SD = 29.7). Of note, just over 2% (12/506) of respondents found research use of DBS "completely unacceptable" across all scenarios, and nearly 9% (45/506) found it "completely acceptable" across all scenarios. We also conducted an Analysis of Variance (ANOVA) to determine whether demographic characteristics impacted acceptability ratings. We did not find any statistically significant effects of respondent race, respondent ethnicity, child and respondent sex, child and respondent age, or parents' awareness of newborn screening (all Fs<3.0, all ps>.09, ns).

[Table 2 about here.]

For the conjoint analysis, data for 432 out of a total of 506 respondents were usable. Of the unusable data (N=74), 45 respondents indicated all scenarios were acceptable, 12 respondents indicated all were unacceptable, 8 indicated all in the mid-range of the response scale, and 9 failed to respond to all scenarios. Conjoint analysis requires that all survey items have responses and that there be some variability among the responses (to denote preferences).

Table 3 contains the fit indices generated by the conjoint analysis. The model showed strong fit with a high Pearson's R by indicating that the predictive conjoint analysis model strongly fits the responses to the eight experimental survey items. The Kendall's tau for holdouts also suggests that the fit between the actual ratings provided by respondents on the eight experimental survey items and the predicted ratings generated by the four holdout model validation items is very strong. The duplicated survey item showed a strong correlation of .87, indicating high response reliability.

[Table 3 about here.]

Figure 2 presents the importance scores. Consent was the most important factor in respondents' ratings of acceptability of the scenarios (average importance score of 64.9). This was followed by identity linkage (average importance score of 19.4), and lastly, the affiliation of the researcher using the DBS (average importance score of 14.6).

[Figure 2 about here.]

Analyses of the part-worth utility values for each of the factors' levels indicate that, focusing on the issue of consent, respondents find it unacceptable (i.e., do not prefer) when consent is not sought for research use of their child's DBS (utility = -25.3). They prefer that their consent be sought for each and every study that would use their child's DBS (utility = 20), followed by their consent being sought one time for all studies that would use their child's DBS (5.3). Regarding the issue of identity linkage, respondents prefer that their child's identity not be linked to his or her DBS (utility = 4.5), compared to when their child's identity would be linked (utility = -4.5). Finally, respondents prefer that the research using their child's DBS be conducted by university researchers (utility = 1.3), compared to researchers from a drug company (utility = -1.3). These part-worth utilities are presented in Figure 3.

[Figure 3 about here.]

DISCUSSION

This predominantly minority, low-income study sample overwhelmingly conveyed that parents do not find acceptable (i.e., do not prefer) situations in which their consent would not be sought in order to use their children's DBS in research. They preferred that their consent be sought for every use of the child's DBS. Though not as strong an influence on attitudes, respondents preferred that the child's identity not be linked to the DBS and the research be conducted by university researchers. Most respondents report that they recall their children

receiving the heelstick at birth, but over a third does not know what newborn screening is. This echoes calls to educate parents about newborn screening practices[33].

This work contributes to the body of literature on parent preferences concerning the research use of their child's biomaterial. Specifically, this research echoes previous work conducted in Michigan highlighting the emphasis parents place on providing their consent to research use of their children's spots[21], but we have quantified the high level of importance parents place on consent relative to other concerns. This work also echoes public attitude assessments surrounding the research use of DBS, in which the opportunity to provide one's permission emerges as a critical theme in both qualitative and quantitative inquiry[24,25].

Limitations and Future Directions

Although this study contributes to the understanding of parents' preferences concerning the research use of their children's biomaterial, several limitations should be noted. First, we only studied three factors that have emerged as important in previous literature (consent, identity linkage, and researcher affiliation), though many others may impact parents' attitudes surrounding the research use of DBS[21-25].

Another limitation is that respondents rated the acceptability of hypothetical scenarios. 'Real' requests to use their children's DBS could produce different results. It is also possible that respondents, especially when completing the survey in a busy, clinic setting, may not have fully understood the information conveyed in the 13 hypothetical scenarios. No respondents in our study sample, nor any of the 15 respondents to the pilot test, indicated that they had difficulty understanding the information being conveyed in the scenarios or the general information presented by the RA regarding newborn screening or blood spot storage. We used the terms "newborn screening" and "heelstick" to gauge participants' awareness and recall of newborn

screening. Some individuals, however, might be more likely to use terms like PKU or sickle cell screening. Further, some respondents' participation was interrupted by being called back for an appointment (N=26, 5% of 506), and there is the possibility that this could have imparted some response bias.

Although not demographically representative of Indiana parents, which are predominantly White[34], this study sample captures individuals who could potentially be asked to provide consent about research use of their children's DBS. Moreover, the importance to parents of providing consent for DBS research use aligns with previous findings, based on data collected from demographically different respondents[21,25].

Lastly, we did not collect data specifically asking about prematurity, neonatal intensive care unit admissions, or other health conditions present during infancy. Presence of health conditions could impact the likelihood of and/or parental recall of newborn screening.

Future directions could build upon this work by eliciting parental preferences for the timing and structure of the consent process. For example, do they prefer an "opt-in" or "opt-out" structure? Do they prefer to be asked at the time of the screening or prior to the baby's arrival? Moreover, future studies could focus on the situations in which identity linkage or consent waivers might be permissible, as well as parental preferences for being informed of study results involving their child's DBS.

CONCLUSIONS

Policymakers, those leading biobanking initiatives, and researchers using minors' biomaterial in their work might benefit from taking parents' preferences into consideration when developing protocols for the use of children's biomaterial, especially consent policies. Though logistic problems with seeking consent for DBS research use might seem overwhelming, such

consent is not necessarily unattainable[35]. Public preferences are critically important for policymakers to keep in mind when developing governance policies; it is also imperative that policymakers understand that preferences can vary depending on the parameters of DBS use. For example, if the DBS are being used in research identifying treatable illnesses, perhaps parents *would* want their children's identities linked to their specimens so that they could be notified.

Further, democratic deliberation approaches that have been employed in biobanking-related research[36,37] and involve intensive education efforts and ongoing discourse between citizens and those responsible for policymaking, might render different sentiments from parents regarding use of their children's biomaterial. However, many individuals from whom consent would be sought for DBS use would not likely be able to participate in long-term, ongoing, intensive education and dialogue about biomaterial use. This study's objective was to measure attitudes from these individuals not undergoing a democratic deliberation process.

Lastly, states that have already implemented governance policies on this topic, especially if it does not involve seeking consent from parents for use of DBS, might benefit from a re-evaluation of such policy.

Figure Legends

Figure 1. Survey item wording. Researcher affiliation, identity linkage, and consent type were varied across the 13 scenarios (see Table 2 for listing of variable combinations reflected in each scenario).

Figure 2. Importance scores depicting extent to which these factors accounted for scenario acceptability ratings.

Figure 3. Part-worth utilities values conveying preferences for the attribute levels within each factor. Values below the baseline of 0 indicated non-preferred (unacceptable) attribute levels and values above the baseline of 0 indicate preferred (acceptable) attribute levels.

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Table 1: Characteristics of parent respondents and the referent child.

		Mean	Min	Max
Ages	Parent Age	32	18	63
	Referent Child Age	5.7	0	17
	Characteristic	**N**		**% (of N responding)**
Parent Race (N=487)	Black or African American	298		61
	White	169		35
	Other	20		4
Parent Ethnicity (N=452)	Hispanic/Latino	19		4
	Not Hispanic/Latino	433		96
Referent Child's Health Care Payer Source (N=483)	Public (Medicaid)	300		62
	Child Covered by Parent's Policy	172		36
	Other/Uninsured	11		2
Parent Income Level (N=486)	Less than $10,000	149		31
	$10,000-$25,000	130		27
	$25,000-$50,000	88		18
	$50,000-$75,000	31		6
	More than $75,000	88		18
Parent Political Affiliation (N=473)	Democrat	250		53
	Independent	156		33
	Republican	67		14
Parent Political Views (N=468)	Very Liberal	131		28
	Moderate	221		47
	Conservative	116		25
Does Parent Consider Self Spiritual or Religious (N=494)	Yes	325		66
	No	54		11
	Somewhat	115		23
Respondent Relationship to Referent Child (N=501)	Mother	419		84
	Father	72		14
	Other	10		2

Table 2: Average acceptability ratings of survey scenarios.

Scenario	Researcher Affiliation	Identity Linkage	Consent	Mean Acceptability Rating	SD
1	University	N	Repeated	78	29.7
2*	Drug Co.	N	Repeated	76.4	31.4
3*	University	Y	Repeated	69.4	35.1
4	Drug Co.	Y	Repeated	68	35.7
5*	University	N	One-time	67.4	36.1
6	Drug Co.	N	One-time	61.4	37.3
7	University	Y	One-time	59.3	38.2
8**	University	Y	One-time	59	38.7
9*	Drug Co.	Y	One-time	57.7	38.7
10	University	N	No	38.2	39.9
11	Drug Co.	N	No	34.4	39.8
12	University	Y	No	30.4	37.3
13	Drug Co.	Y	No	28.6	36.4

Scenario ratings were provided on a scale anchored at 0 (completely unacceptable) to 100 (completely acceptable) in increments of 10. Order presentation was randomized between participants in five different survey versions.

**Denotes that scenario was a holdout item for model validation.*

***Scenario #8 duplicated scenario #7 as index of response reliability.*

Table 3: Indices of conjoint analysis model fit.

Fit Index	Value	Significance
Pearson's R for Conjoint Model	.998	$p < .001$
Kendall's tau for Conjoint Model	.929	$p = .001$
Kendall's tau for Conjoint Model Holdouts	1.000	$p = .02$
Pearson's R for Duplicated Scenario Validation Check	.874	$p < .001$

Figure 1: Survey item wording. Researcher affiliation, identity linkage, and consent type were varied across the 13 scenarios (see Table 2 for listing of variable combinations reflected in each scenario).

*Researchers at a university [**drug company**] would like to perform a study using the blood spots collected from your child when they were born. Your child's identity would be linked to their spots but would be kept private [**would not be linked to their spots**]. The researchers would ask you for your permission one time in order to use your child's blood spots for all studies they plan to do [**would ask you for your permission separately each and every time they wanted to use your child's blood spots in a different study;** would use your child's blood spots without asking you for your permission].*

Figure 2. Importance scores depicting extent to which these factors accounted for scenario acceptability ratings.

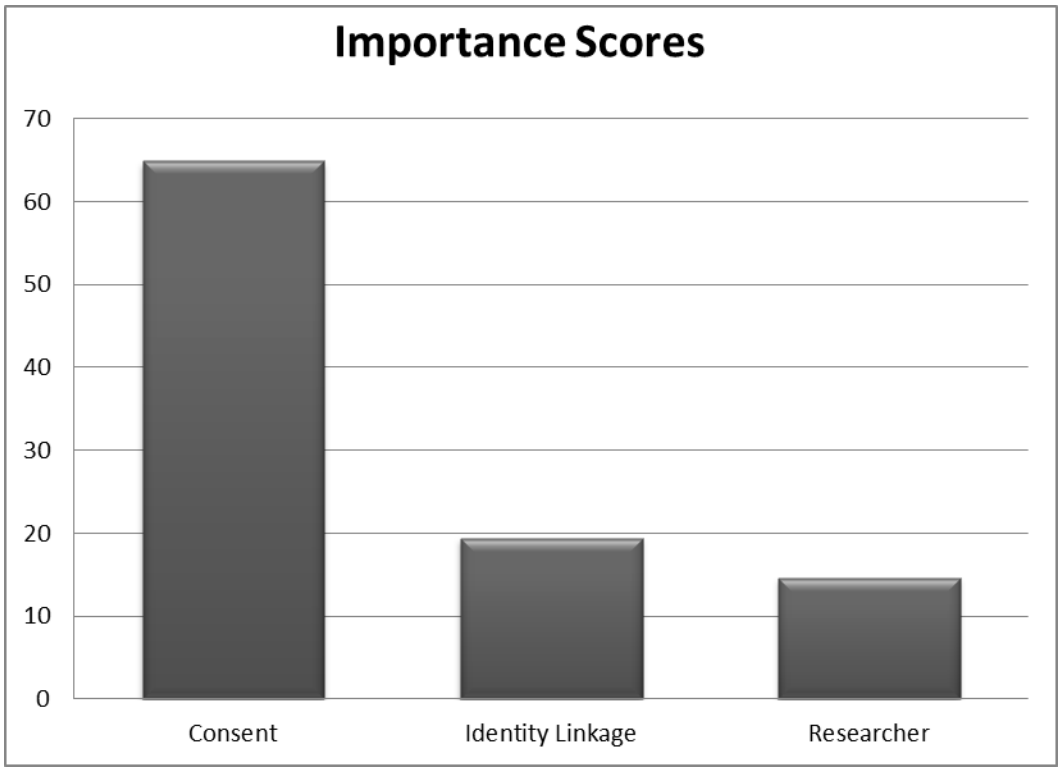

Figure 3.Part-worth utilities values conveying preferences for the attribute levels within each factor. Values below the baseline of 0 indicated non-preferred (unacceptable) attribute levels and values above the baseline of 0 indicate preferred (acceptable) attribute levels.